

REVIEW ARTICLE

Shaken Baby Syndrome

A Common Variant of Non-Accidental Head Injury in Infants

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SUMMARY

Background: Recent cases of child abuse reported in the media have underlined the importance of unambiguous diagnosis and appropriate action. Failure to recognize abuse may have severe consequences. Abuse of infants often leaves few external signs of injury and therefore merits special diligence, especially in the case of non-accidental head injury, which has high morbidity and mortality.

Methods: Selective literature review including an overview over national and international recommendations.

Results: Shaken baby syndrome is a common manifestation of non-accidental head injury in infancy. In Germany, there are an estimated 100 to 200 cases annually. The characteristic findings are diffuse encephalopathy and subdural and retinal hemorrhage in the absence of an adequate explanation. The mortality can be as high as 30%, and up to 70% of survivors suffer long-term impairment. Assessment of suspected child abuse requires meticulous documentation in order to preserve evidence as well as radiological, ophthalmological, laboratory, and forensic investigations.

Conclusions: The correct diagnosis of shaken baby syndrome requires understanding of the underlying pathophysiology. Assessment of suspected child abuse necessitates painstaking clinical examination with careful documentation of the findings. A multidisciplinary approach is indicated. Continuation, expansion, and evaluation of existing preventive measures in Germany is required.

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Key words: head injury, brain damage, child abuse, pediatric care, forensic medicine

Child abuse can take the form of physical cruelty, neglect, or emotional or sexual abuse (1). Abuse-related craniocerebral trauma or non-accidental head injury (NAHI) accounts for only a small proportion of all child abuse, but is conspicuously over-represented in the first year of life (2). NAHI is the most frequent non-natural cause of death in infancy (3), and the most common cause of death overall between the ages of 6 and 12 months (4). The clinical spectrum ranges from trivial bruising to severe trauma with fatal outcome. The shaken baby syndrome (SBS) is a common form of NAHI in which the victim is held by the torso or the extremities and violently shaken, causing abrupt uncontrolled head movements with a marked rotatory component. Clinically, SBS is characterized by signs of severe diffuse cerebral trauma, i.e., acute encephalopathy, subdural hemorrhage, and retinal hemorrhage, occasionally accompanied by various combinations of metaphyseal fractures or rib fractures. The explanation provided for the injuries is frequently inadequate or inconsistent (3, 5).

Some basic aspects of SBS, e.g., with regard to the biomechanics and the cause of death, have not been fully clarified (e1–e3). Recent, sometimes emotionally tinged scientific controversy over the diagnostic value of subdural hemorrhage in infants has further increased the uncertainty (e3–e5). Against the backdrop of recent well-publicized cases of fatal child abuse, the aim of this article is to depict the current state of knowledge on SBS. The scientific literature is reviewed and the recommendations of national and international expert panels are summarized.

Historical review

Abuse and killing of children has long been recognized as a phenomenon occurring throughout human history. However, the medical discovery of SBS did not begin until the second half of the 20th century (6, e6–e8). In 1946 the American pediatrician John Caffey described infants with fractures of the long bones and subdural hemorrhage (e9). Caffey suspected that this constellation could have arisen from unnoticed or concealed accidents, but did not yet realize that what he was seeing was actually a characteristic syndrome following abuse. In 1962 Henry Kempe published his observations on the "battered child syndrome," the first comprehensive scientific article on the topic of child abuse (7). In 1971 the British neurosurgeon Norman Guthkelch described

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TABLE 1

Selected differential diagnoses of SBS

| Constellation | Remarks, references |
|---|---|
| Accidental craniocerebral trauma | Serious accidents rare in infancy, often with SDH and fractures, very seldom with RH (e97–e102) |
| Perinatal | SDH found in 8%, RH in 34% of newborns; generally resorbed by four weeks with no ill effects (e108, e109) |
| Aneurysm/AVM | Rare as cause of bleeding in infancy; exclusion by imaging |
| Arachnoid cyst/ external hydrocephalus in BESS | SDH possible following trivial trauma, exceptionally associated with RH; diagnosis by imaging, possibly only after observation for some time (e110, e111) |
| Meningoencephalitis | Postinfectious hygroma possible, exceptionally associated with RH; diagnosis by imaging, CSF, laboratory tests (18) |
| Coagulopathies | SDH and RH possible; diagnosis by laboratory tests |
| Terson syndrome | Very rare in infancy, in contrast to adulthood (e112) |
| Glutaraciduria type I | Exceptionally associated with SDH and RH; disease course generally features characteristic crises; usually already known (part of neonatal screening) (e71, e72, e76–e78) |
| Galactosemia | Intraocular bleeding described in exceptional cases; generally characteristic clinical picture featuring hepatosplenomegaly, jaundice, sepsis, cataract; usually already known (e113) |
| Osteogenesis imperfecta type I/type IV | Atypical fractures possible; generally characteristic clinical picture with positive family history, blue sclera, wormian bones; molecular genetic diagnosis in exceptional cases (e73) |
| Menkes syndrome | SDH described in isolated cases (e75); characteristic clinical picture featuring microcephaly and typical trichopathy (kinky hair disease); diagnosis by laboratory tests |
| Increased intrathoracic/ intravasal pressure | RH very exceptionally after resuscitation, fits; not after vomiting or coughing (25, e114, e115) |

SBS, shaken baby syndrome; SDH, subdural hemorrhage; RH, retinal hemorrhage; AVM, arteriovenous malformation; BESS, benign enlargement of the subarachnoid space

two infants with subdural hemorrhage but no signs of external injury; as the cause, he suspected an acceleration-deceleration mechanism ("whiplash injury") (e10). In 1972 followed Caffey's seminal study on SBS, in which he was the first to link a shaking event with a constellation, henceforth recognized as typical, of subdural hemorrhage, retinal hemorrhage, and fractures of the long bones (8). Caffey is therefore regarded—while acknowledging the important work done by Kempe, Guthkelch, and others—as the first to describe SBS.

Epidemiology

Child abuse is a global problem (9, 10, e11–e15). No detailed data on the killing of infants or on the incidence of SBS are available for Germany, neither from official registries nor in the scientific literature.

Analysis of German police statistics for the year 2006 indicates an annual incidence of around 30 (recorded) cases of abuse and three homicidal deaths per 100 000 children under the age of 6 years (e16, e17). In a multicenter study of sudden infant death (SIDS) in Germany, autopsy revealed SBS as the cause of death in almost every 50th case diagnosed as SIDS (e18). Further improvement in the accuracy of the data can be expected from the ongoing Survey of Rare Pediatric Diseases in Germany (ESPED, Erhebungseinheit für Seltene Pädia-

trische Erkrankungen in Deutschland; www.esped.uni-duesseldorf.de [in German]).

Extrapolation of the data from the few epidemiological studies—principally from English-speaking countries and reporting rates of between 15 and 30 per 100 000 children under the age of 1 year (e19–e23)—yields an estimated incidence of 100 to 200 cases per year in Germany (e16). SBS is the most severe form of abuse in infancy and the most frequent non-natural cause of death; more than two-thirds of all fatal cases of child abuse occur in this age group (2). Over 90% of all serious intracranial injuries in infancy result from abuse (e24).

Causes and pathophysiology

Child abuse is a multifactorial phenomenon. The risk factors for SBS include low socioeconomic status, disability of the child, violent tendencies, and alcohol or drug abuse within the family (11, 12, e25–e28). However, SBS occurs in all social strata (12). A typical constellation is that of a baby that "cries all the time" with young, overstressed parents whose repeated efforts to pacify the child end in failure and who have a low frustration threshold and poor control of impulses (e29, e30). In the case of inadequate social resources, a stress situation may grow ever more acute until it ends with

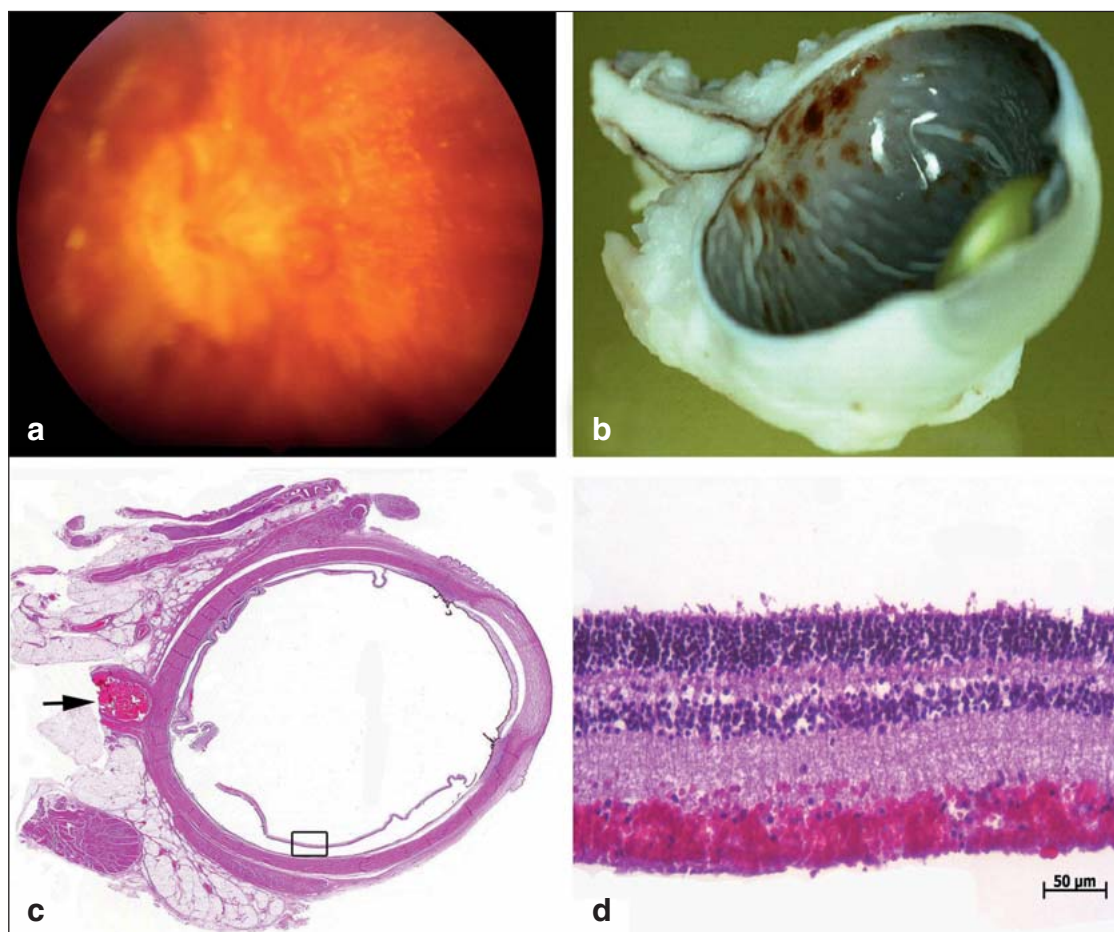


Figure 1: Ophthalmological findings in shaken baby syndrome.
a) Funduscopy (from the collection of Professor Alex Levin, Ophthalmology and Vision Sciences, Hospital for Sick Children, University of Toronto, Canada) and b) macroscopic view of surgical specimen with typical "flame-like" retinal hemorrhages c) Histological ocular specimen showing bleeding in the optic nerve sheath (arrow) and retinal hemorrhages d) Enlargement of the area marked in c)

loss of control and shaking of the child (4). In surveys carried out in the USA, 50% to 75% of teenagers and young adults stated they did not know that shaking is dangerous and between 2.6% and 4.4% of the parents of children under 2 years of age reported they had shaken their child at least once (e31–e33). The equivalent figure for parents in Indian city slums was 42% (e32). Recently initiated public awareness campaigns in the USA have shown promising early results (e34). Efforts to raise awareness of the particular dangers of shaking and of the help available have also begun in Germany, specifically in Hamburg and Lower Saxony (leaflet for young parents in Hamburg: "Hilfe! Mein Baby hört nicht auf zu schreien," www.hag-gesundheit.de/documents/schuette_130.pdf [in German]). However, these measures have not yet been evaluated. It is the responsibility of the courts to make the sometimes extremely difficult judgment of whether there was any intention to kill or harm the child. Nevertheless, many perpetrators are probably well aware of the dangers (e35). The perpetrator is most commonly the child's father or the mother's new partner, less frequently the mother or a female babysitter (3, e36, e37).

A number of anatomical features make infants particularly vulnerable to acceleration-deceleration events with a marked rotatory component, which typically

occur on shaking (2, 13–15, e38, e39). The head is large in relation to the rest of the body and is not yet adequately supported and controlled by the weak, immature neck musculature (14, e40). The result is vigorous movements of the various intracranial compartments relative to one another, e.g., between the skull and dura on the one hand and the cerebral surface on the other, or between the white matter and the gray matter. Although many details remain unclear, the overwhelming majority of investigators agree that the resulting shear forces are responsible for subdural hemorrhages and diffuse brain damage (3, 14–15, e38–e44). "Simple" shaking without impact suffices to produce the full picture of SBS with or without fatal outcome, but the energy resulting from an abrupt deceleration through impact is certainly higher and thus leads to more severe trauma (shaken impact syndrome) (e45–e49).

Contrary to previous opinion, it is no longer thought that subdural hemorrhages are of any relevance with regard to the prognosis or the cause of death (14, 15). Subdural hemorrhages typically form a thin film rarely more than 2 to 3 mL in volume and thus do not constitute a space-occupying lesion (14, e46, e50). Rather, they can be seen as a diagnostic marker for the severe acceleration-deceleration trauma that is typical in shaking. One important harmful factor is central apnea,

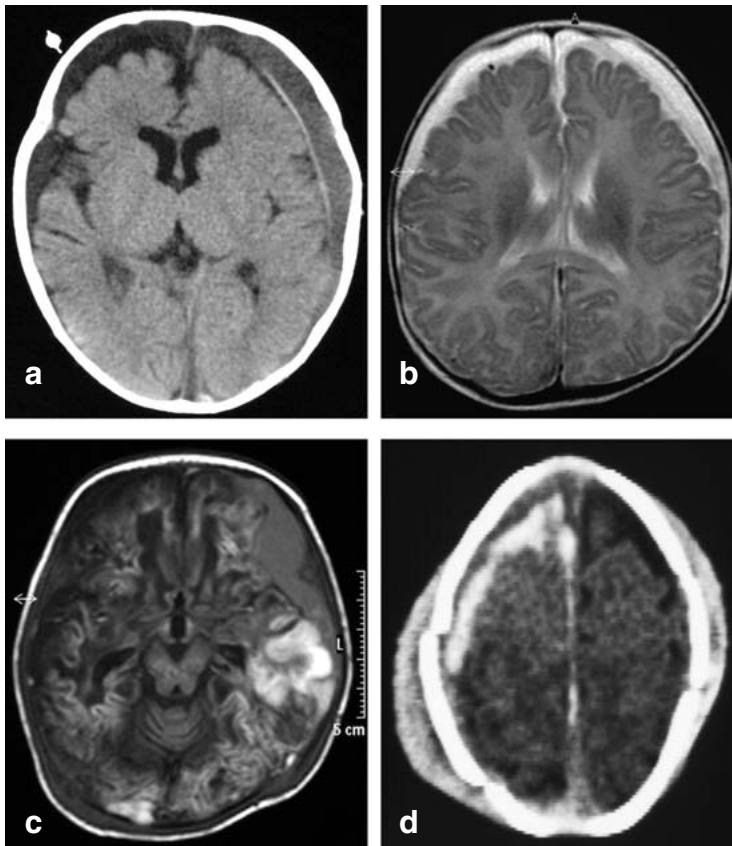


Figure 2:

Radiological findings in shaken baby syndrome (SBS)

- a) Cerebral computer tomography (CCT) in a male infant with suspicion of SBS: bilateral subdural hemorrhages of variable density
- b) T2-weighted cerebral magnetic resonance imaging (cMRI) of a male infant with suspicion of SBS in the past: bilateral hygromas
- c) cMRI of a male infant around 1 month after admitted shaking: severe residual defects, status post diffuse parenchymal injury and left intracerebral hemorrhage and subdural hemorrhage
- d) CCT of female infant with SBS in combination with severe contact trauma ("shaken impact"): subdural hemorrhage, dislocated cranial fracture, bilateral soft tissue trauma (from the collection of Professor Gabriele Benz-Bohm, University Hospital of Cologne)

triggered by a sudden marked extension of the medulla oblongata on shaking (16). Neuropathological investigation has revealed signs of corresponding focal damage in the lower brainstem (e51, e52). Even if a long-lasting episode of apnea is not immediately fatal, the resulting hypoxia causes cerebral edema with increased intracranial pressure and thus reduced cerebral blood flow, leading to a vicious circle of increasing cerebral hypoxia. The end result in such cases—depending on the delay before initiation of emergency treatment—is either protracted brain death or prolonged survival with serious deficits (e53–e55). Moreover, the shear forces that act on the immature brain during shaking result in traumatic diffuse axonal injury (DAI) (e51, e52), which also participates in the development of cerebral edema (16, e56). For forensic purposes, it is important that in SBS—following from biomechanical and pathophysiological principles—impairment of cerebral function, or symptoms, begin immediately after the shaking event. In other words, a shaken infant who displays severe neurological symptoms at a later stage is unlikely not to have shown signs of injury straight after shaking. Furthermore, the accounts given by many confessed perpetrators describe occurrence of the symptoms immediately following non-trivial shaking (e48, e57–e59.).

Clinical findings and diagnosis

In less severe cases the symptoms are often unspecific (e.g., vomiting, sleepiness) and can be misinterpreted as enteritis, infection, irritability, or failure to thrive (4, e60). The patients are usually unconscious and bradycardic, floppy, or cramping when brought to the hospital, with no coherent explanation why they are in such a condition. The carers commonly make statements such as: "I found her like this," "suddenly turned blue while I was feeding him," or "fell off the couch three days ago." Appraisal of SBS can be complicated by the statement that the child was found apparently dead and shaken in an attempt to revive it. In this regard, the American Academy of Pediatrics has stated that shaking sufficient to cause SBS is so violent that an observer would

BOX 1

Procedure in the case of suspected child abuse*1

History

- How it happened / social history / previous medical history

Physical examination

- Whole-body examination (always)
- External injuries (distribution / pattern / form / color / extent)

Documentation

- Written description / sketch / photographs (scale!)

Diagnostic procedures

- Laboratory tests (blood count, liver function, coagulation, CSF)
- Ophthalmologist (funduscopy)
- Radiologist (CCT / MRI, x-ray skeletal screening)

Interdisciplinary conference and planning involving:

- Treating primary care physician
- Hospital
- Youth Welfare Office / social worker
- Psychologist / psychiatrist
- Forensic medicine
- Parents (if willing to cooperate)

*1After careful consideration of legal implications or in the case of danger to life or physical integrity, inform police and/or Youth Welfare Office/family court

recognize it as immediately life-threatening (5). Patients with SBS have a poorer prognosis than victims of serious accidents; this can be attributed in particular to the differences in mechanism of injury and the frequent delay in taking the child to a doctor (e61–e64). Mortality rates of 13% to 36% are reported in the literature; the proportion of survivors with lasting impairments varies according to study design between 62% and 96% (e65–e70). Laboratory tests can demonstrate or exclude a number of differential diagnoses that may exceptionally mimic symptoms of SBS (*table 1*) (17, e71–e78).

Retinal hemorrhage can be demonstrated in 50 to 100% of SBS patients. They are usually extensive, displaying bilateral symmetry, and located at the posterior pole or in the vicinity of the ora serrata (*figure 1*) (18, 19, e79–e82). Their mechanism of origin has not been finally clarified; however, analogous to the origin of subdural hemorrhage, it is assumed that the acceleration-deceleration causes relative movements of the vitreous body on the one hand and the retina and vessels on the other (e82–e84). Most studies support a link between the extent of retinal hemorrhage and the severity of the trauma suffered or the prognosis (e79–e81, e84–e87). Investigation and documentation by an ophthalmologist are therefore necessary (e88).

Supplementary radiological examinations (*figure 2*) are of great importance for evaluation, monitoring, and documentation (20–22, e89). Although cerebral magnetic resonance imaging (cMRI) is more sensitive than cranial computed tomography (CCT) in the detection of diffuse parenchymal lesions and also superior in estimating how much time has elapsed since injury, for logistic reasons CCT is more commonly performed in the acute situation (20). Moreover, CCT has the advantages of (a) shorter examination times in clinically unstable patients and (b) simpler operation when the clinical symptoms require scanning of the thorax, abdomen, or spine (22, e90). cMRI should then be performed within a few days, and again after one to two months to monitor progress. T2, T2*, FLAIR, and diffusion weighted sequences should be obtained in all planes; contrast enhancement is usually unnecessary (22). Conventional and Doppler sonography can be used for initial orientation. All infants in whom abuse is suspected should undergo radiographic bone screening, possibly preceded by skeletal scintigraphy, for detection of peripheral fractures; skull x-ray is recommended only if CCT is unavailable (4, 5, 12, 23). SBS-related subdural hemorrhage, which can be demonstrated in over 90% of cases, are usually located bilaterally over the convexity and in the interhemispheric fissure and posterior cranial fossa (e50, e91). Epidural hemorrhages or contusions, however, are rare (15).

Differential diagnosis and recommended course of action

Experience of case appraisals and literature reports are instructive in showing that SBS is frequently not diagnosed or, if correctly diagnosed, doesn't lead to appropriate action (e60, e92, e93). The most common

BOX 2

Child abuse: important legal provisions

BGB*1 § 1631: Content and limits of care of persons

(2) Children have a right to non-violent upbringing. Corporal punishments, emotional injuries and other degrading measures are prohibited.

SGB*2 VIII § 8a: Duty of protection to children at risk

(1) If the Youth Welfare Office has credible information indicating a risk to the welfare of a child or adolescent, it must assess the risk in cooperation with specialist personnel. The persons having care or custody and the child/adolescent should be involved in consultation, unless the effective protection of the child/adolescent would thereby be endangered. If the Youth Welfare Office considers assistance to be appropriate and necessary to avert the risk, it must offer this assistance to the persons having care or custody of the child/adolescent.

Offences against life (homicide)*3

§ 211 StGB*4: Murder under specific aggravating circumstances
§ 212 and 213: Murder, Murder under mitigating circumstances
§ 222: Negligent manslaughter

Offences against the person*3

§ 223 StGB: Causing bodily harm
§ 224 and 226: Causing bodily harm by dangerous means,
Causing grievous bodily harm
§ 225: Abuse of position of trust
§ 227: Infliction of bodily harm causing death
§ 229: Causing bodily harm by negligence

StGB § 203: Violation of private secrets (duty of confidentiality)

(1) Whosoever unlawfully discloses a secret of another, in particular, a secret which belongs to the sphere of personal privacy or a business or trade secret, which was confided to or otherwise made known to him in his capacity as a
1. physician, dentist, veterinarian, pharmacist or member of another healthcare profession which requires state-regulated education for engaging in the profession or to use the professional title shall be liable to imprisonment of not more than one year or a fine.

StGB § 34: Necessity

A person who, faced with an imminent danger to life, limb, freedom, honor, property or another legal interest which cannot otherwise be averted, commits an act to avert the danger from himself or another, does not act unlawfully, if, upon weighing the conflicting interests, in particular the affected legal interests and the degree of the danger facing them, the protected interest substantially outweighs the one interfered with. This shall apply only if and to the extent that the act committed is an adequate means to avert the danger.

RiStBV*5 No. 235: Child abuse

(1) Even anonymous and confidential information is always followed up by the district attorney; In the course of securing evidence he pays particular attention to observation of § 81c para. 3 clause 3 StPO*6.
(2) In the case of child abuse there is always a special public interest in the prosecution of the offence (§ 230 para. 1 clause 1 StGB). Conversion to a private prosecution according to § 374 StPO is generally not indicated.
(3) If socio-pedagogical, family therapy, or other supportive measures have been initiated and seem to promise success, the public interest in prosecuting the offence can lapse.

*1 Bürgerliches Gesetzbuch (German Civil Code); *2 Sozialgesetzbuch (German Social Code); *3 These offences are prosecuted ex officio even if no complaint is filed or a complaint is filed and then withdrawn; *4 Strafgesetzbuch (German Criminal Code); *5 Richtlinien für das Strafverfahren und das Bußgeldverfahren (Guidelines for Criminal Proceedings and Summary Proceedings); *6 Strafprozeßordnung (German Code of Criminal Procedure)

Key messages

- The shaken baby syndrome is a frequent and characteristic form of non-accidental head injury in infants.
- The clinical manifestations include signs of severe diffuse brain damage and subdural and retinal hemorrhage; the explanation given is often inadequate.
- The prognosis is extremely poor, with mortality of around 30%; in survivors, the literature reports the rate of long-term impairment at 62% to 96%.
- The brain damage and bleeding are caused by severe cranial acceleration-deceleration trauma with a marked rotatory component arising from violent shaking of an infant or young child that has no control over its head movements.
- Following promising early efforts at prevention in the USA, expansion and evaluation of similar measures, e.g., public awareness campaigns, are called for in Germany.

reasons are lack of specialist knowledge or diagnostic uncertainty (e92, e94). The situation is often complicated by the parents' vehement/aggressive denial of abuse. In many fatal cases there are signs of previous abuse (e95), underlining the importance of reliable diagnosis and/or appropriate action.

Subdural hemorrhage or retinal hemorrhage can be found in infants in situations other than abuse (*table 1*); in the great majority of cases, however, the combination of subdural hemorrhage and retinal hemorrhage is a result of SBS (5, e19–e21, e96). The vast majority of the numerous accidents that happen to infants every day do not result in serious injury (e97–e100). Severe accidental head injuries are extremely rare in this age group and usually the result of falls from great heights or high-speed vehicle accidents (e101, e102). The proportion of patients with retinal hemorrhage after severe accidental head injury is under 3% (24, 25, e103–e107).

The recommended procedure in the case of suspected child abuse is outlined in *box 1*. The precise course of action should be adapted according to the individual situation. Early contact with forensic specialists is advisable; many forensic institutes and some children's hospitals have dedicated pediatric units on 24-hour standby. The welfare of the child must always have absolute priority. On the other hand, imprudent action in the case of false suspicion can do lasting harm to the welfare of the family. Hospital admission of an SBS patient is indicated not only on medical grounds but also to avoid the risk of further abuse. There is no general answer to the question of whether and when the law enforcement authorities should be informed; careful consideration is required in each individual case. If, after such consideration, there is the impression of serious, unavertable danger to the child's life or physical integrity, §34 of the German Criminal Code (StGB) permits the vow of confidentiality to be broken (*box 2*). However, in the clinical context there is no obligation to notify the authorities. If it is decided to discharge the child to the care of the family, close medical monitoring, including any siblings, and close cooperation of the parents with the Youth Welfare Office are absolutely

essential ("help, not punishment"). In such cases the law enforcement authorities may abstain from prosecution (*box 2*). In the case of death, however, any signs of abuse found post mortem must be reported to the police or district attorney. A forensic autopsy with supplementary neuropathological investigation should then be ordered by the authorities—experience shows that this is not a matter of course and has not always happened in the past.

Conflict of interest statement

The authors declare that no conflict of interest exists according to the guidelines of the International Committee of Medical Journal Editors.

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